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Recurrent intrascleral cyst after strabismus surgery

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Abstract ● Background: Intrascleral epithelial inclusion cysts have been described after ocular trauma, scleral buckling and strabismus surgery. They are usually small, asymptomatic and located anteriorly. ● Case report: The clinical hisotry of a 9-year-old girl who developed a huge epi- and retrobulbar intrascleral cyst in both the upper and the lower nasal quadrants after multiple strabismus operations is described. ● Results: Surgical resection of the cyst wall was twice unsuccessful. Drainage of the cyst, followed by the injection of tetracycline (30 mg/ml) in the cyst site, led to complete recovery. ● Conclusion: Recurrent posteriorly located intrascleral cyst can occur after strabismus surgery. Therapy consisting of drainage combined with intrascleral administration of tetracycline solution (30 mg/ml) to induce sclerodesis appeared to be effective during a 2-year follow-up period.

Introduction

Epithelial inclusion cysts occasionally develop after trauma, strabismus surgery or scleral buckling surgery. After strabismus surgery, they are usually small and have an anterior episcleral location close to, or even in, an operated muscle [3, 6, 9]. After scleral buckling surgery they can some-times develop in the posterior part of the orbit, and can cause problems due to increasing size [4]. Reports of intrascleral implantation cysts are few in number and suggest a traumatic aetiology [1, 8].

The case we describe here is the first reported recurrent symptomatic extensive intrascleral cyst after strabismus surgery. We suggest a new treatment.

Case report

A 9-year-old girl was referred to our department because of a painless decrease in visual acuity (VA) in her right eye over a 3-week period. She had a history of repeated strabismus surgery, performed elsewhere 3, 7 and 8 years previously. The superior, medial and lateral rectus muscles had been recessed and the inferior oblique muscle partially disinserted. Her VA was normal after these procedures in both eyes.

On admission, the patient's best corrected VA was 20/100 in the right and 20/20 in the left eye. External examination showed mild protrusion (Hertel 17-80-15 mm) and esotropia of the right eye. An elastic mass was palpated through the lids on the upper part of the globe between the 9 o'clock and 2 o'clock positions. Slit-lamp examination of both eyes was normal. Fundus examination of the diseased eye showed a huge indentation of the upper quadrants up to the disc, protruding over the macula (Fig. 1). The left eye was normal. Ultrasonography revealed a well-outlined epi- and retrobulbar process, connected to the globe, in three quadrants, protruding mainly anteriorly, with a maximal antero-posterior diameter of 15 mm (Fig. 2). While no internal reflectivity was observed, this finding was suggestive of a cystic lesion.

Because of the treat to VA, surgical intervention was judged to be necessary. After puncture of the cyst, clear watery fluid was drained and the external wall of the cyst was resected as far posteriorly as possible. One day after surgery a dramatic improvement of the fundus aspect was observed (Fig. 3), and 4 weeks later VA had increased to 20/25. Histological examination confirmed the diagnosis of an intrascleral cyst with a scleral outer surface and an inner surface lined with nonkeratinised multilayered squamous epithelium (Fig. 4). Goblet cells were not present in the epithelium. A small amount of scar tissue and inflammatory cell reaction around remnants of suture material was seen.

Nine months after surgery, the patient was readmitted with the same complaints and clinical findings as at the initial referral.

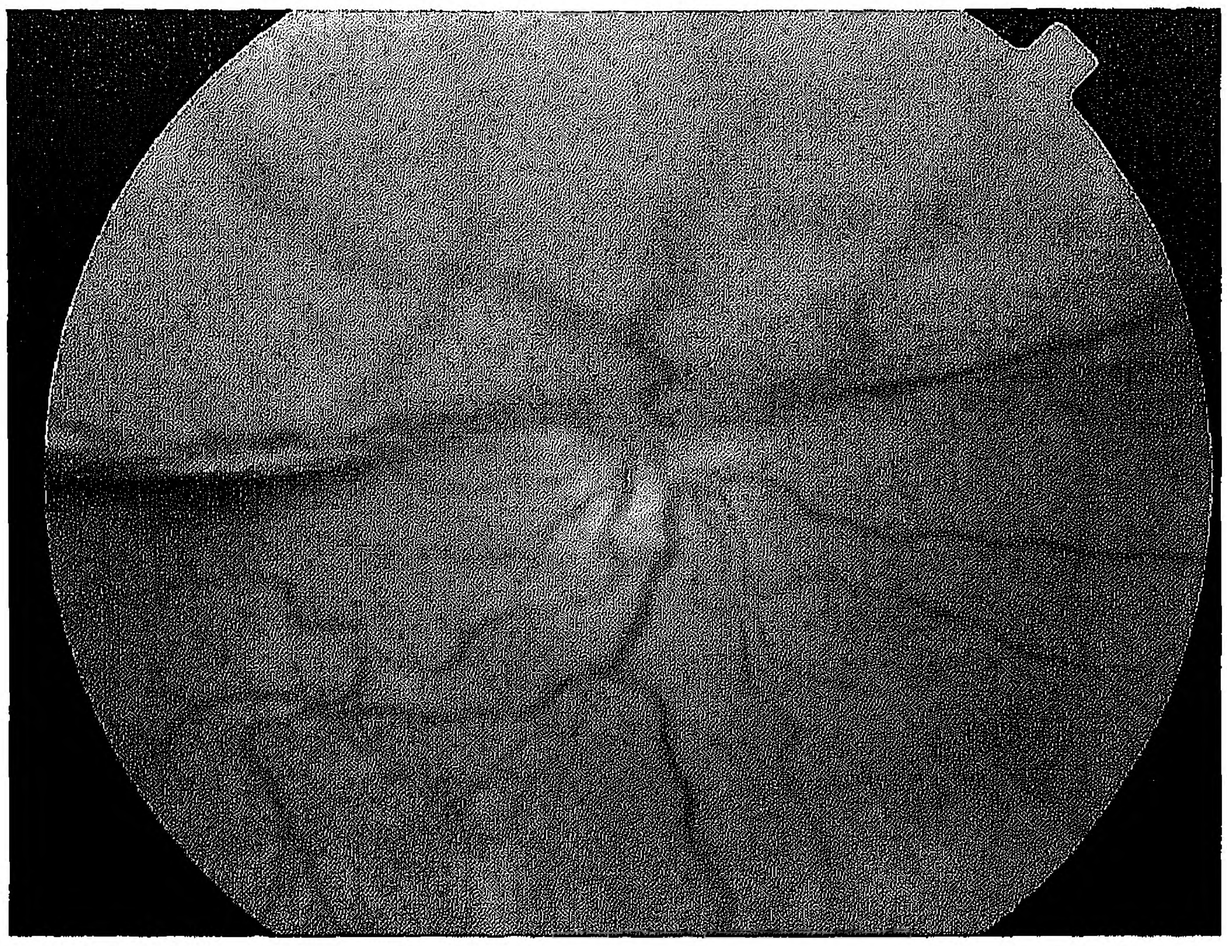

Fig. 1 Appearance of the fundus, showing a large indentation of the superior quadrants of the globe

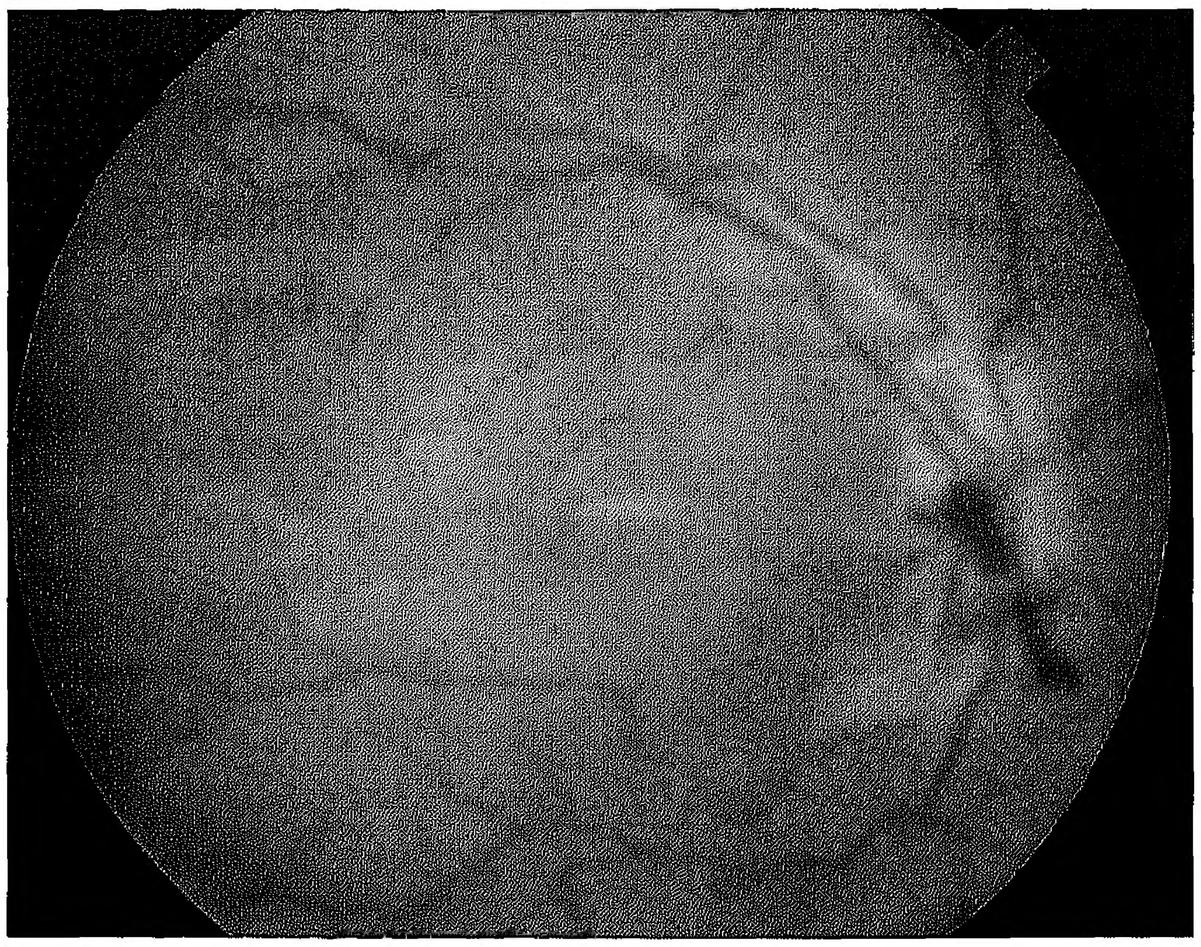

Fig. 3 Appearance of the fundus aspect 1 day after the first operation, showing papilloedema and haemorrhages on the disc and the retina

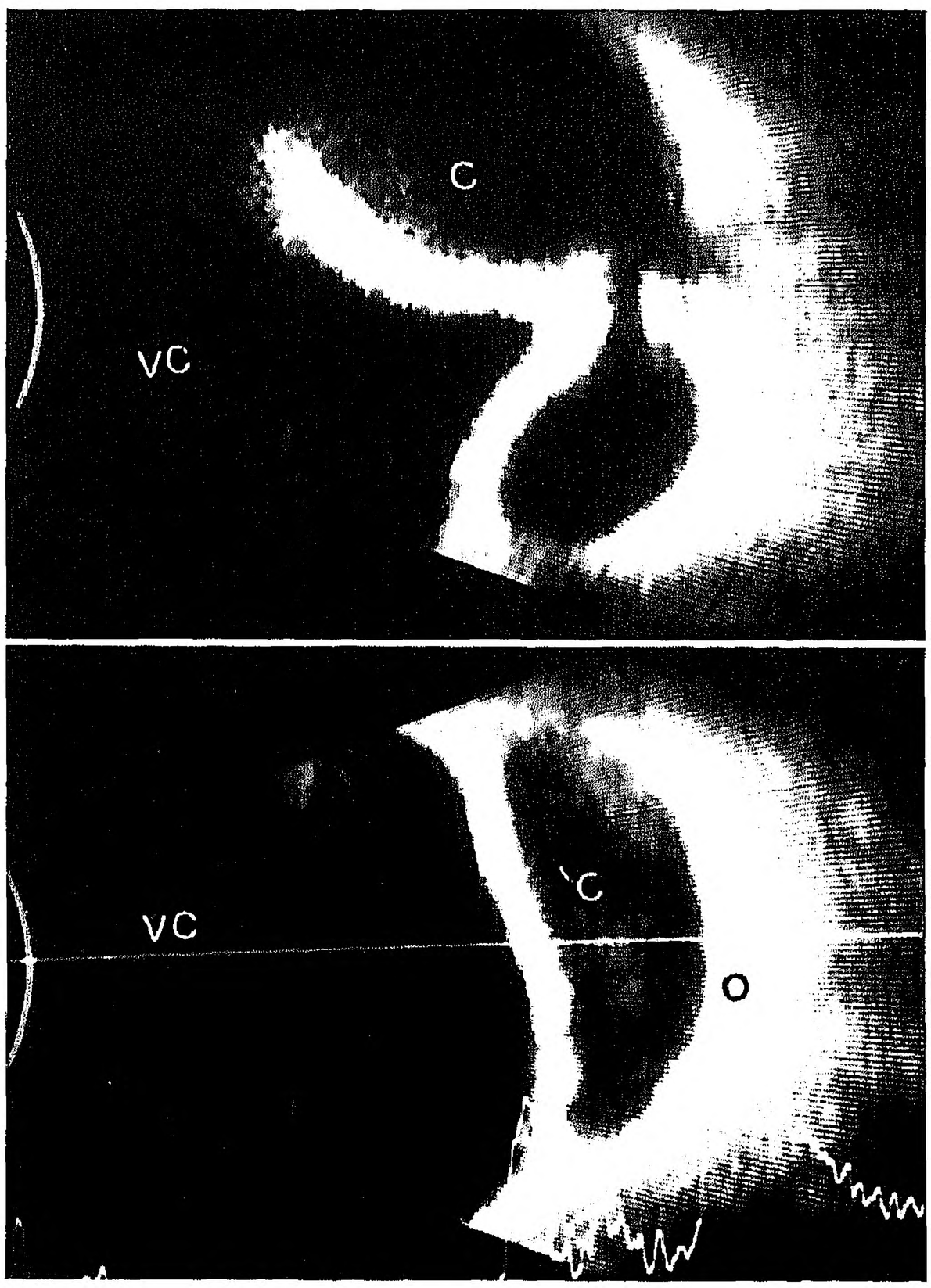

Fig. 2 Ultrasonography of the right eye. *Top and bottom left:* B-mode scans obtained with the transducer on the globe, showing a round hypoechoic lesion. *Top and bottom right:* Immesion B-mode scans showing the strong indentation and the location around the optic nerve. (*VC* vitreous cavity, *O* orbital fat, *C* cyst, *ON* optic nerve)

Fig. 4 Histologic section of the cyst wall, showing the internal surface of the cyst lined with nonkeratinised multilayered squamous epithelium without goblet cells adjacent to scleral stroma

Recurrence of the cyst was ultrasonographically confirmed. An even more radical excision of the cyst wall was performed. Four months after this operation, however, the cyst reappeared for the third time. We decided on another approach.

After drainage of the cyst, 0.4 ml of a concentrated tetracycline solution (30 mg/ml) was injected between the cyst walls to induce sclerodesis. No further surgery was performed. Four months after this procedure, the VA was restored to normal. Two years later there were no signs of recurrence of the cyst, either clinically or ultrasonographically.

Discussion

During strabismus surgery, conjunctival epithelium may be snagged in the suture material and pulled into the scleral tunnels when the sutures are knotted [6].

Intrascleral epithelial inclusion cysts are rare, probably because scleral tissue is a poor culture medium for the epithelial cells [1]. In our patient, four muscles were ''attacked'' seven times in three surgical procedures. Each intervention increases the chance of an inclusion cyst. The mainly anteriorly directed extension of the cyst in this patient suggests an inclusion in the deeper scleral layers. The origin of the clear fluid in the cyst remains unexplained because of the absence of goblet cells. Two recurrences despite careful and extensive surgery were unexpected.

Chemical pleurodesis with tetracycline has been shown to be successful in the treatment of recurrent pneumothorax [2, 5, 7]. We decided, therefore, to try an analogous approach in this case, in the hope that the acidic tetracycline solution might produce sclerodesis, which then did actually occur. During a 2-year follow-up period no signs of recurrence or toxicity were observed on or in the treated eye.

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